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No Burp Syndrome or Retrograde Cricopharyngeus Dysfunction in a Filipina College Student: A Case Report

ABSTRACT

Objective: To report a case of retrograde cricopharyngeal dysfunction (R-CPD) and the use of botulinum toxin injection for its diagnosis and therapy.

Methods:

Design: Case Report

Setting: Tertiary Private University Hospital

Patient: An 18-year-old female college student who presented with inability to burp, associated with awkward gurgling noises from the chest, abdominal discomfort and excessive flatulence.

Results: Our patient gained the ability to belch and the relief of associated excessive flatulence and awkward gurgling noises from the chest, after cricopharyngeal muscle Botulinum toxin injection.

Conclusion: Botulinum toxin injection may serve as both a diagnostic and therapeutic intervention for R-CPD.

Keywords: *retrograde cricopharyngeal dysfunction; R-CPD; botulinum toxin injection; cricopharyngeus muscle*

The cricopharyngeus muscle (CPM) is an important structure that functions as the upper esophageal sphincter (UES). The ability to burp, mainly governed by relaxation of the CPM or UES, is a reflex that helps the human body remove excessive gas accumulating in the upper gastrointestinal tract and may serve as a protective mechanism to expel entities disrupting the upper gastrointestinal tract.[1] The inability to belch may lead to debilitating aerodigestive symptoms which were first described by Kahrilas *et al.* in 1987,[2] as a belch reflex dysfunction causing severe chest pain. This condition was eventually named Retrograde Cricopharyngeal

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Dysfunction (R-CPD) by Bastian *et al.* in 2019,[3] as a syndrome consisting of the inability to belch accompanied by gurgling noises from the chest, abdominal discomfort and excessive flatulence. R-CPD may be debilitating and socially crippling which affects quality of life. Along with its introduction, Bastian and Smithson[3] demonstrated the use of botulinum toxin injection as both a diagnostic and therapeutic intervention for R-CPD. Several articles have recently been published, illustrating that such a syndrome is gradually being recognized.[3-11] Here we present a case of an 18-year-old female college student with R-CPD symptoms who was managed with botulinum toxin injection.

CASE REPORT

An 18-year-old female college student claimed that she did not have the ability to eructate since she was a child with undetermined onset. This inability to belch presented with episodes of abdominal bloating, gurgling sounds from her chest and neck and excessive flatulence. These symptoms were triggered by intake of spicy or sweetened food and carbonated drinks, and were prominent after a heavy meal, which caused great discomfort. No medical consultations were previously made, and she also had an otherwise unremarkable birth and medical history.

It was around three years prior to consultation that the abovementioned symptoms became persistent and already affected her personally and socially, in terms of bonding with her family and friends on occasions that would involve eating. Being an only child, her parents were highly involved in her unusual situation and were driven to research if such a condition even existed. With the help of the internet and social media, the parents came across a syndrome called Retrograde Cricopharyngeal Dysfunction through a *YouTube* video by Dr. Bastian of the Bastian Voice Institute. This gave them the idea of having the patient consult with an ear, nose and throat (ENT) specialist.

A few months prior to consult, she finally had an initial check-up with an ENT specialist and was assessed to have unremarkable physical examination findings. Having noted her symptoms involving the aerodigestive tract, she was referred to a laryngologist. The laryngologist ruled out possible pathologies that may have caused her inability to belch, through a battery of tests such as flexible laryngoscopy to check for possible aspirations or obstructions and modified barium swallow to evaluate dysfunctions in swallowing. *Figure 1* shows the unremarkable flexible laryngoscopy of the patient. There was no edema nor hyperemia noted in the patient's arytenoids and epiglottis. Her true vocal cord mobility was good with adequate glottic opening and complete glottic closure. The pyriform sinus is not obliterated and no mass or obstruction was seen. Moreover, modified barium swallow studies showed that she had intact swallowing.

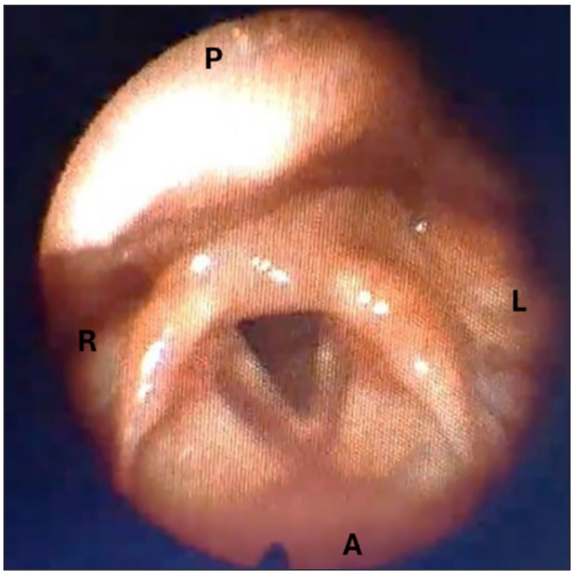

Figure 1. Unremarkable laryngoscopy, (A) Anterior, (P) Posterior, (R) Right, (L) Left.

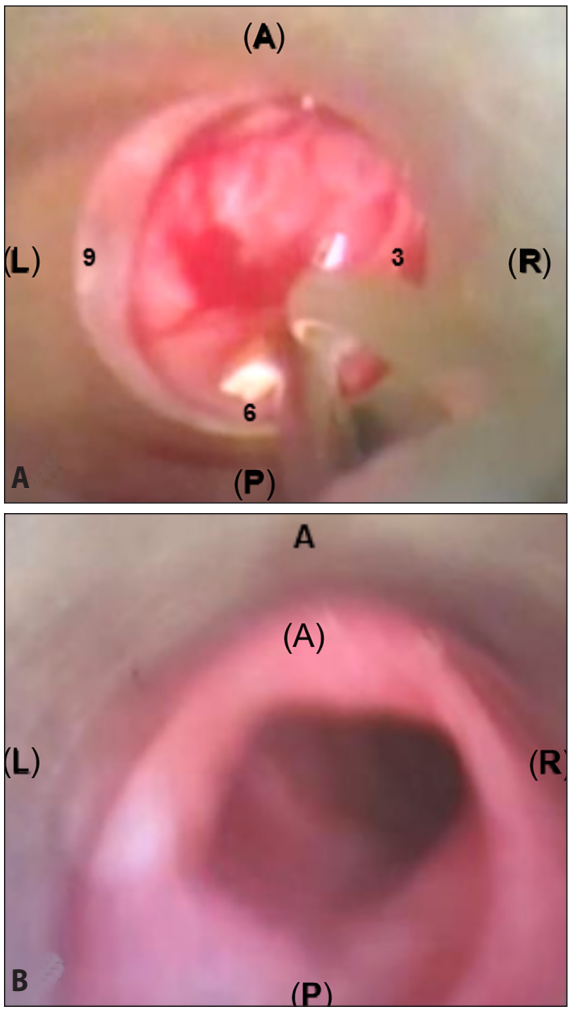

Figure 2. A. Botulinum toxin injection sites at 3, 6 and 9 o'clock positions; **B.** smooth upper esophagus: (A) Anterior, (P) Posterior, (R) Right, (L) Left.

At this point, her diagnosis remained undetermined, with R-CPD as the only consideration. To solidify the diagnosis, the patient was referred to another laryngologist from our institution, who appraised her for botulinum toxin injection.

She underwent direct laryngoscopy with botulinum toxin injection of the cricopharyngeus muscle (CPM) under general anesthesia. A total of 45 units was injected at the junction of the esophageal mucosa and muscle, 15 units at 3 quadrants particularly at the 3, 6 and 9 o'clock positions. Intraoperatively, the rigid laryngoscope was advanced further at the post-cricoid area. The upper esophageal sphincter was exposed and identified. *Figure 2A* demonstrates injection of botulinum toxin into the cricopharyngeus muscle, specifically at the junction of the mucosa and the muscle, at 3, 6 and 9 o'clock positions, while *Figure 2B* shows the upper esophagus which was smooth with no lesions noted.

After surgery, our patient was advised soft diet for the next few days and prescribed with anti-reflux medication. Follow-up was conducted at one week, one month, three months and six months. As early as three days after the procedure, our patient experienced her first "small burp." She described it as small gasses of air which frequently and unpredictively come out of her mouth. This was accompanied by relief of her abdominal discomfort and gurgling noises from the chest. However, with the ability to belch, she also noted globus sensation, dysphagia to solids and acid reflux. These other symptoms eventually resolved after one month. Currently at six months post-surgery, our patient claims to still be able to belch, with no more complains of loud gurgling sounds coming from the chest, and less abdominal discomfort and flatulence. She claims to be able to take in a wider variety of food but still in moderation especially with trigger foods. Along with the resolution of symptoms, she is able to attend social gatherings and engage in social interactions better after having received the treatment. She also claims to enjoy her food experience and became less anxious. Overall, the intervention gained positive results for our patient over six months. We are still for monitoring her for possible relapse and retreatment.

DISCUSSION

The clinical approach to diagnosis for this case involved a proper history and physical examination. Our patient matched all the symptoms present in R-CPD patients including the inability to belch, abdominal discomfort, gurgling noises from the chest and excessive flatulence.[3] Moreover, she experienced social dissociation which is a noted psychological effect of this syndrome.[4] It has been established that most R-CPD patients have a heightened occurrence of psychiatric symptoms in relation to their inability to burp which affects their quality of life.[4] She had also undergone a battery of tests to investigate her aerodigestive tract which all yielded normal results. As described in several studies, numerous R-CPD patients underwent extensive testing such as esophagoscopy, barium studies, and esophageal manometry which yielded unremarkable results and connote that these may play little role in its diagnosis.[3,5,7,10] Nevertheless, one study noted that manometry and barium studies may help in ruling out certain disease entities such as achalasia.[6] Having R-CPD as the diagnosis of exclusion, our patient was offered botulinum toxin injection to confirm R-CPD and to manage the disease as well.

Based on systematic reviews, the mean dose of Botox injection given to R-CPD patients which demonstrated therapeutic effect is 66 units and reported success rates range between 88.2–95% with a preferred dose of botulinum toxin from 25U–100U.[7,8] Although there was a study where pediatric R-CPD patients were given a total of 25-50 units of Botox injection with high success rate for treatment response.[9] Taking into account that in this case we had an 18-year-old young patient, a total of 45 units of Botox injection was given which yielded satisfactory results even at 6 months.

Based on our patient's preference, this procedure was performed via direct suspension laryngoscopy under general anesthesia. Although Botox injection can be done through either direct suspension laryngoscopy under general anesthesia or trans-cricothyroid membrane puncture, the former has high requirements in terms of equipment and technology while the latter has high requirements in terms of skill and precision.[11]

Immediately after the procedure, our patient claimed to have unpredictable and frequent "micro burps." This was followed by the relief of her abdominal discomfort and gurgling noises from the chest. Aside from resolution of her symptoms, she also claimed to have globus sensation and dysphagia, which are expected postoperative discomfort as effect of injection. Based on studies, mild transient dysphagia is the most common.[10-11] However, these mild discomforts eventually resolve over the course of 1-2 weeks. In this case, her symptoms of globus sensation and dysphagia eventually resolved after 4 weeks.

Previous R-CPD patients had to interact with their newfound ability to belch to control initiation or timing.[3] During patient surveillance in this case, the nature and timing of burp onset, alterations in swallowing and reflux symptoms were monitored. Our patient was also advised to maintain a soft diet and was instructed to eat slowly to manage the swallowing changes happening within the first two to four weeks. In terms of patient satisfaction, she claimed to have better social interactions and social bonding. Studies had demonstrated that R-CPD patients who underwent Botox injection had substantial improvement in the quality of life, describing it as "life changing."[3,4,11]

In terms of effect longevity, a retrospective study done by Hoesli *et*

al.,[11] have shown 80% of R-CPD cases maintained a satisfactory ability to burp until 26 months post-treatment, while 20 % of them lost the ability to burp within minimum of 4 months. These patients eventually underwent several retreatment sessions depending on longevity of therapeutic effect. In the case of our patient, constant surveillance is still advised to document the duration of the treatment response and to provide necessary retreatment if indicated.

As Bastian and Smithson[3] described, botulinum toxin injection was more or less a therapeutic diagnosis. Our patient did not receive a correct diagnosis or helpful treatment until she was matched with a specific syndrome and treated with botulinum toxin. Based on this case report as well as other related studies,[3-11] diagnosis seems to be validated by the combined clinical assessment given the history of unremarkable laboratory studies, and treatment with botulinum toxin. Lastly, this case serves as a reminder that there are patients who are information driven. It is the obligation of health care providers to keep up with the continuously evolving collection of information which may be important in treatment outcomes, which is the improvement of the patient's quality of life.

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